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Running Title: Expression of Pain in Children: Autism Spectrum Disorder

The Expression of Pain in Children:
An Autism Spectrum Disorder Perspective

By

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ABSTRACT

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ADVISOR: Cay Anderson-Hanley, PhD.

Pain sensitivity in children with Autism Spectrum Disorders (ASD) have been shown to vary greatly in the literature. Previous work by Rattaz et al. used venepuncture to display slower recovery in ASD youth. Later, Duerden et al. used the Cold Pressor Test (CPT) to conclude that ASD youth demonstrate a profile of decreased thermal sensitivity.

In the current study, four normative males and one ASD male participated in the Cold Pressor Test. The ASD youth was also given the Gilliam Autism Rating Scale (GARS-3) to verify ASD tendencies. Prior to the CPT, all participants were asked to place their dominant, unclenched hand to wrist in a warm water bath for two minutes, then the same hand was placed in the CPT. They were asked to provide pain intensity ratings, using the Visual Analog Scale (VAS) at the initiation of pain, every ten seconds, and upon withdrawal of the hand. Baseline vitals were also obtained prior to and following both the warm water bath and CPT. Upon completion, video recordings of the participants were reviewed for facial actions by two raters using the Child Facial Coding System (CFCS) to assess pain. The ASD child was found to provide lower VAS scores compared to his normative peers, yet both groups showed the same number of facial actions during the two CPT iterations.

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INTRODUCTION

Autism was first discussed by Psychiatrist Leo Kanner in 1943. He described 11 children whose development and behavior were so strikingly different from their typically developing counterparts, including “autistic aloneness” and insistence on sameness. Following this, Hans Asperger in 1944 identified in 4 boys a pattern of behavior and abilities that included “a lack of empathy, little ability to form friendships, one-sided conversations, intense absorption in a special interest, and clumsy movements” (Frith, 1991).

Autism Spectrum Disorder (ASD) is defined as a serious neurodevelopmental disorder, impairing a child’s ability to communicate and interact with others. Here, the term “Spectrum” refers to the wide range of symptoms and severity. ASD has been diagnosed based on deficits in social communication and repetitive or restricted behavioral routines (Moore, 2015). As of March 2014, the Centers for Disease Control has identified 1 in 68 American children, with 1 in 42 boys versus 1 in 189 girls having ASD (CDC, 2016). There is no singular cause for this disorder. While there have been numerous genetic mutations associated with Autism, most cases appear to be multifactorial with autism risk genes and environmental factors, such as advanced parental age, maternal diabetes or infection during pregnancy, premature birth with very low birth weight, and birth complications including oxygen deprivation influencing early brain development (Autism Speaks). The brainstem has been suggested as a key area for symptoms associated with Autism as well as for sensory modulation. Hardan et al. in 2008, researched the involvement of the thalamus in the sensory abnormalities of Autism using MRI and proton spectroscopy (Klintwall et al., 2011). A year later, Jou et al. in 2009, used MRI scans coupled with a Sensory Profile Questionnaire, showing a relationship between decreased brain-stem grey matter and oral sensory sensitivity (Klintwall et al., 2011). A more recent study from Columbia

Medical Center, noted that a pruning defect during a child's development would lead to excess synapses causing Autism symptomology (Pedersen, 2015).

A child with Autism is unable to be definitively diagnosed by a physician until 18 to 24 months. However, caretakers can look for their children not reaching "Developmental Milestones," such as failure to reciprocate sounds, smiles, or other facial expressions by 9 months, no babbling or pointing by 12 months, or a loss of babbling, speech, or social skills at any age. This type of pattern is referred to as "regression" (National Autism Association). This spectrum encompasses Autistic Disorder ("classic autism"), Rhett syndrome, Childhood Disintegrative Disorder, Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS), and Asperger Syndrome. ASD has been associated with intellectual disability, difficulties in motor coordination and attention, and physical health issues such as sleep and gastrointestinal disturbances. Many individuals with ASD excel in visual skills, art, music, and math. While about 40% have intellectual disability (IQ less than 70), many have normal to above average intelligence, and also, while 25% are nonverbal, they are able to learn how to communicate through other means (Autism Speaks).

Sensory differences have also been noted to be qualitatively different in individuals with ASD. However, while these symptoms have been associated with ASD, there is question of whether this symptomology is a core component of ASD deficits, or rather a co-morbid phenomenon. For this facet to be a core feature, it has to be universal, meaning that almost all individuals with ASD would need to have this symptom. It needs to be unique, such that individuals with non-ASD diagnoses do not have this same symptom. Finally, it has to be specific, meaning that it differs from the other core symptoms within the ASD diagnosis. Ben-Sasson and colleagues examined the sensory modulation disorders (SMD) that afflict individuals

with ASD. They reviewed over-sensitivity, under-sensitivity, and seeking, with the latter describing the craving of, and interest in sensory experiences. Previous literature has suggested that several factors contribute to sensory modulation symptoms in individuals with ASD: chronological age (CA), mental age (MA), and severity of ASD. Review through meta-analysis revealed that there was an increase in over-responsivity and seeking up to age 6-9 years, with a decrease following this age group. However, there was no consistent trajectory for under-responsivity amongst age groups. Due to this, there is a need to study non-socially related under-responsivity symptoms in order to gain insight into whether under-responsivity dominance in those with ASD is due to their inherent desire to be socially withdrawn, or whether these symptoms have a greater sensory basis (Ben-Sasson et al., 2009).

Professional literature has also suggested that individuals with ASD are usually insensitive to painful stimuli. However, some researchers have presented a possible hypersensitivity facet to Autism, but this is not as often endorsed. It has been noted that hypersensitive individuals will demonstrate an abnormally sensitive behavioral response to stimuli, which are deemed by others to be benign. Also, hypersensitive individuals are susceptible to sensory overload, leading to heightened anxiety and a response that leads the individual to withdraw from the environment. Contrastingly, hyposensitive individuals have decreased awareness or are completely unaware of stimuli in their environment, which can lead them to harm. Pain has been defined by the International Association for the Study of Pain as ‘an unpleasant sensory and emotional experience associated with actual or potential tissue damage’ (IASP, 1994). One’s sensitivity for pain is composed of not only somatic sensory perception but also of subjective emotional reaction. Evolutionarily, pain is a key signal for preserving one’s self (Yasuda et al., 2016). Self-reported pain by high-functioning ASD

individuals and their parents have been studied. Children with ASD and a non-ASD group rated their hypothetical pain when shown pictures depicting common childhood situations (i.e. scraping a knee on the sidewalk), and also provided a rating for the amount of pain they would expect to feel using the Faces Pain Scale. Parents also gave pain ratings for what they expected their child to feel. Results showed that ASD individuals did not differ significantly from their non-ASD peers on pain ratings, nor did parents (Bandstra et al., 2012). While pain in this study was not induced, and was only hypothetical, it adds further discrepancy to the literature, since it shows that high-functioning ASD youth can understand pain.

The research in Autism and pain is particularly salient for three reasons. First, under Autism Spectrum Disorder in the DSM-V, the sensory aspect criterion of ‘apparent indifference to pain/heat/cold’ has been returned (American Psychiatric Association, 2013). Second, individuals with ASD are more likely to experience painful episodes throughout their lives, due to the greater prevalence of comorbid conditions associated with painful procedures, and the greater amount of likely accidents (National Autism Association). Third, a majority of studies with children on the Autism Spectrum have focused on processing of other senses, such as auditory and visual processing (National Autism Association). Thus, this possible pain facet necessitates consideration of how individuals with ASD communicate pain differently to those around them, which leads to alterations in pain expression and response.

Early Studies

Initial studies in the field concerning pain and children on the Autism Spectrum, involved only one child on the Spectrum. Beginning with Mahler’s 1952 account of a child, who was presented an electric torch and car lighter, both of which were placed in his mouth, burning his lips, yet he did not cry. Mahler deduced from this that children on the Spectrum are

hyposensitive (Moore, 2015). Rothenberg in 1960 further bolstered this assertion by noting a child, Johnny who did not feel pain even upon being hit hard (Moore, 2015). Wing in 1976 discussed a child, Sally, who would play outside in the snow without clothes (Moore, 2015). More recently, Gilberg and Coleman in 2000 observed a child who would place his hand on the stove and was only made aware of this by the smell of burning skin (Moore, 2015). While these are all case studies with no normative comparison group, they anecdotally provide support for the claim of hyposensitivity among children on the Autism Spectrum.

Other studies recruited numerous children on the Autism Spectrum in an effort to quantify the hyposensitivity to pain. In 1971, Kolvin found that 25% of 48 ASD participants were unresponsive to pain, however there was no comparison to a normative population and a clear methodology was not provided (Moore, 2015). Gilberg et al. in 1985, found that 55% of 20 ASD children were hyposensitive to pain based on reactivity to injections, bruises and pain caused by childhood injury, but again, there was no comparative group and non-standard measures of pain (Moore, 2015).

The assertion of hyposensitivity among the ASD population was eventually contradicted through Bursch et al. presentation in 2004 of two cases of pain in ASD adults. ‘Tony’ had long-term headaches and chronic abdominal pain, and ‘Greg’ also had chronic abdominal pain. However, there was an incident where Greg grabbed a hot frying pan but failed to express pain as someone in the normative population would (Moore, 2015). Tony, however, always expressed his pain. While there still was no normative population comparison, this observation of the two ASD participants did suggest the possibility of not only hyposensitivity, but also hypersensitivity.

Experimental Examination

In 2010, Bird et al. gave ASD and normative adults a square pulse waveform to their hand to measure electrocutaneous pain thresholds. Participants were to indicate their pain on a 10-point scale. Results showed that ASD adults did not differ in pain threshold compared to the normative group (Moore, 2015). Using ASD and normative adults as well as adolescents, in 2013, Fan et al. studied pressure pain thresholds. They applied pressure to the finger or hand, and then participants reported their pain rating. It was found that ASD participants had lower pain thresholds, or more sensitive to pain than their normative counterparts (Moore, 2015).

Medical Procedure

With the need for a normative comparison group, Nader (2004) recruited 3-7 year olds with Autism and non-impaired children, who were matched for gender. These participants were given an intravenous injection, during which video recording was employed with 10 second segments being later evaluated by two raters, using the Child Facial Coding System (CFCS), and Observational Scale of Behavioral Distress (OSBD). Following completion of the study, parents were administered the Faces Pain Scale (FPS) to report on the child's pain, and prior to the study, the parents completed the Non-Communicating Children's Pain Checklist (NCCPC), remarking on prior pain reactions of the child. The children with Autism displayed a significant facial pain reaction to the venepuncture method compared to their non-impaired peers, and there was a lack of concordance between the parental reports of pain and the observed pain responses of the children (Nader et al., 2004). This finding of greater facial activity by ASD children, illuminated the possibility that this population may not be hyposensitive to pain, and instead they may have a problem accurately communicating their pain to others.

Another study dealing with Autism Spectrum children undergoing venepuncture, but dealing more with the impact of facial activity on observer's ratings of pain intensity was

conducted by Messmer et al. (2008). Facial activity raters were provided information that the ASD children may have potential pain hypersensitivity, pain hyposensitivity, or no difference in pain sensitivity from their normative peers. Participants were then presented an information booklet on Autism, then given video clips of the ASD children experiencing venepuncture, and these had already been coded by the CFCS. However, participants were asked to rate pain intensity of the child based on the VAS. Results showed that children who received lower scores on the CFCS were deemed to be experiencing lower intensity pain. This study showed that raters VAS scores, being influenced by the facial pain activity of the ASD children, indicated that the child's experience of pain was being communicated in some way.

In another study utilizing venepuncture, Tordjman et al. in 2009 compared age and sex matched children on the Autism Spectrum to their normative peers. While the ASD children were less responsive during venepuncture, there was increased self-injurious behavior following conclusion of the procedure. This increased behavior is suggestive of increased distress, presenting the possibility that ASD children do not react in the same way to pain as their normative peers but that they do experience pain. More biologically, Tordjman et al. (2009) showed that ASD children displayed elevated heart rate and plasma β -endorphin levels (physiological reactivity) when in pain.

Similar to Tordjman et al. (2009), Rattaz et al. (2013) noted the behavioral and physiological reactions of ASD children. However, these children were compared not only to typically developing children, but also to children with intellectual disabilities. During the venepuncture, all participants were videotaped and had their heart rate recorded. Following the procedure, a different investigator used the CFCS to code facial actions and the NCCPC to code behavioral responses every 10 seconds, as well as obtaining the heart rate every 10 seconds.

While there were no significant differences between the groups for facial, behavioral and physiological reactions, behavioral reactions remained high in the ASD children after the end of the venepuncture. This shows that ASD children recover more slowly, which contrasts the findings of Tordjman et al. (2009).

The Cold Pressor Model /thermal models

Modir and Wallace (2010) reviewed the effectiveness of the cold pressor (CP) as a reliable model to test pain, with participants submerging their hands to forearms into icy water, assessing the onset to pain, pain intensity, and tolerance. All of these measures utilized the VAS pain rating scale. The researchers further elaborated on the impacts of the CP, discussing initially a cold pain sensation to be felt, followed by a radiating deep, dull aching pain. The CP effectively produces a reproducible acute and noxious cold pain stimulus to the participant. Their review of the CP disclosed the varying outcomes, which can be the products of different CP temperatures and gender differences, as well as the exclusion criteria for the CP. Their meta-analysis revealed that participants are tested in a quiet and comfortable room without distractions, their dominant arm and hand temperature are equilibrated by submersion into a 35°C warm water bath for 2 minutes, and they are then asked to submerge the same hand into a continuously circulating 1°C cold water bath until the pain proves intolerable. During this cold water bath, participants provide VAS pain ratings and tolerance time is measured, with the study being ended at 120 seconds.

In 2008, Cascio et al. (Moore, 2015) compared adults with high-functioning Autism to their normative counterparts on cold and hot thresholds. A thermode increasing 1.5°C/s was administered until participants noted the feeling of pain. Results showed that ASD individuals had lower pain thresholds, indicating a hypersensitivity to pain. A study conducted in 2015 by

Duerden et al. used a computer-controlled deliver system of thermal stimuli. For the warm detection threshold (WDT) and cold detection threshold (CDT), participants pressed a button when they first felt a warm or cool sensation. They were also given a heat pain threshold (HPT) and cold pain threshold (CPT). Here also, they pressed a button when they felt heat or cold pain. It was found that ASD individuals had significantly higher WDTs and lower CDTs than the normative group. However, while trends were seen in the ASD group towards increased HPT and decreased CPT, there was no significant difference from the normative group.

To understand the ethical concerns of using the CPT with children, Birnie et al. (2010), surveyed researchers who have used the CPT in studies, along with children who have participated in these studies and their parents. It is unclear if the CPT meets the criteria for the United States Code of Federal Regulations Part 46 Protection of Human Subjects, describing the procedures which entail a minor increase over minimal risk. Common concerns raised by the CPT is the unnecessary induction of physical pain to children, the level of induced pain, which would cause unnecessary psychological distress, lack of benefit to the participating child, and that the CPT is greater than minimal risk. However, following a CPT study with a child, a majority of researchers received positive feedback from the family. Similarly, when children and their parents were surveyed, the parents rated the experience very positively and children who had participated in more than one CPT rated the experience more positively than those who had completed only one CPT.

In regards to administering the CPT to children, von Baeyer et al. (2005) introduced specific guidelines for this population. The CPT has been used in at least 24 published studies involving children since 1937, and has been administered to more than 1700 children aged 4 to 18 years. To minimize the risk to the participating child, there is exclusion criteria, a maximum

immersion time in place for hand submersion into the CPT, and the child remains in control of the process. While CPT studies for adults regularly use temperatures between 0-5°C, von Baeyer suggests 10°C (Birnie et al., 2010). This temperature has been found to maintain a warm enough water temperature so that the child does not withdraw the hand within a few seconds of submersion, and also does not cause so little pain that all children keep their hands in the CPT until the maximum immersion time. It is also suggested that hand to wrist is sufficient to create pain from the CPT and that dominant or non-dominant hand use does not have an impact. Similar to the review done by Modir and Wallace (2010), von Baeyer et al. (2005) notes a 2-minute hand submersion into 35-37°C warm water to aid in acclimation of the child to the laboratory and study.

The Cold Pressor Test has been shown to increase blood pressure in healthy patients, and heart rate is more contingent upon the individual. In terms of heart rate, it is often the case that an individual will have a sustained increase throughout the CPT, or heart rate will decrease after an initial increase (Mourots et al., 2008). Tassorelli et al. (1995) also elaborated on the CPTs impact on the cardiovascular system, noting a significant increase in blood pressure after two minutes and remaining elevated, whereas heart rate was not significantly different.

Yasuda et al. (2016) delved further into developing a method for evaluating sensory abnormalities in ASD individuals. ASD individuals were sex- and age-matched to a normative group, with objective pain sensitivity as well as subjective pain sensitivity being measured. Objectively, pain was measured following the electrical stimulation, while heat and cold noxious stimuli were presented via a Peltier device. For each measurement, a detection threshold, pain threshold, and pain tolerance were measured. For subjectivity, pain intensity was measured via VAS scores, and the short form of the McGill Pain Questionnaire (SF-MPQ) was used for pain

intensity and type. It was found that the objective perception of pain was not impaired in ASD individuals, but they did exhibit hyposensitive subjective pain sensitivities. Thus, ASD individuals have an impaired cognitive processing mechanism in regards to pain.

The use of the cold pressor test (CPT) for the cold pain threshold facet of the Duerden et al. (2015) study led to the question of reliability. Turk and colleagues stated that the CPT was the most valid method for measuring pain (Birnie et al., 2010). While the CPT is a reliable means of delivering a noxious pain stimulus, disparity in the literature regarding results following administration of the CPT could be due to varying temperatures used. Mitchell et al. (2004) noted that a difference of as little as 2°C could produce a different result. Therefore, during this study's administration of the CPT, the temperature was monitored.

Building on previous research regarding pain expression in children on the Autism Spectrum (Yasuda et al. 2016; Duerden et al. 2015; Rattaz et al. 2013), it was examined whether pain elicited a difference in pain expression between the ASD child and the normative group. Following a warm water bath, all participants were given the CPT. During the CPT, the participant gave visual analog scale (VAS) pain ratings at the onset of pain, every 10 seconds during the CPT, and when the participant withdrew his hand. Besides the subjective measure, a video recording was employed throughout the study to objectively review facial expressions via the child facial coding system (CFCS), following termination of the study.

Hypotheses:

1. It is expected that high functioning children with ASD are not hyposensitive to pain.
2. It is expected that these children express pain differently than their normative counterparts.

METHODS

Participants

The sample (n=5) consisted of one male child with ASD from the community aged 16 years and a normative population of four male children from the community aged 13 years. All participants were Caucasian and from the Capital region. The participants were volunteers who were solicited through fliers in various nearby public locations, Union College campus listserv, and connections among fellow students. Specifically, the ASD children were recruited through Campbell House and local Autism organizations and events, and the normative group was recruited through Shenedahowa and Brown schools. Each potential participant was screened by phone or email for inclusion in the study via questions about past medical history that would prohibit their ability to participate in the cold pressor test (CPT) task. Thus, study risks and benefits were reviewed and all study participants signed assent forms and caretakers signed consent documents approved by the Institutional Review Board at Union College.

Procedures

The study was run for 30 minutes. The child was taken into a separate room for the study with few auditory and visual distractions and only the researcher present, while the caretaker completed the demographics questionnaire and Gilliam Autism Rating Scale, 3rd edition (GARS-3). The participants had their baseline vitals, such as heart rate and blood pressure taken upon entering the lab. The researcher then showed the child how to properly place the hand in the water bath. While standing, the participant placed the dominant, unclenched hand to wrist into a 35°C water bath for 2 minutes to equalize the skin temperature, and this was recorded, along with the vitals. The researcher then informed the participant of the visual analog scale (VAS) pain rating scale and reiterated the child's ability to withdraw the hand at any time during the study. The participant was then asked to submerge the hand into the cold water bath of 10°C

until it proved intolerable. At the initiation of the CPT, the participant was asked to state when there was the onset of pain (threshold) and the time was recorded via stopwatch. Finally, the time at which the participant withdrew the hand was recorded (pain tolerance). Throughout the CPT, VAS ratings of pain intensity were taken at the pain threshold, withdrawal, and every 10 seconds. Since this study employed children, there was a maximum immersion time of 2 minutes. Baseline vitals were taken again. The warm water bath and CPT were administered once more following the 2-minute rest period. Throughout the study, a videotaping system was employed to later review facial actions based on the Child Facial Coding System for 10 second segments. This review was conducted by two raters, ensuring interrater reliability. The GARS-3 was used to note the repetitive behaviors displayed by the child during the study. The VAS pain ratings, CFCS scores, and vitals were interpreted to compare the pain experience between the child with ASD and the normative population.

Measures

Child Facial Coding System (CFCS; Gilbert et al., 1999). This system codes 13 facial actions, and at times codes for intensity (range: 0-2), rather than presence. It was developed for use in children aged 2 to 5 years old but has since been applied to adolescents.

Visual Analog Scale (VAS). This pain rating scale uses 0 to 10. 0 indicates no pain present and 10 indicates an unbearable pain.

Vitals. Heart rate and blood pressure were obtained via a blood pressure cuff that was positioned on the participants left arm upon arrival into the lab, after the warm water bath and before the CPT, and after the CPT.

Gilliam Autism Rating Scale, 3rd edition (GARS-3; James Gilliam, 2014). Items on the GARS-3 are based on the 2013 diagnostic criteria for Autism Spectrum Disorder adopted by the

APA and published in the Diagnostic and Statistical Manual of Mental Disorders, 5th Edition (DSM-5). The instrument consists of 56 clearly stated items describing the characteristic behaviors of persons with Autism. The items are grouped into six subscales: Restrictive, Repetitive Behaviors, Social Interaction, Social Communication, Emotional Responses, Cognitive Style, and Maladaptive Speech. All six subscales have been empirically determined to be valid and sensitive for identification of children with ASD. It is used to assess the severity of Autistic Disorder in the participants for research purposes and to provide a standard diagnostic criterion. The GARS-3 was designed to identify and diagnose Autism in individuals between the ages of 3 and 22 years. The scale aims to differentiate Autism from other conditions with some similarities in presentation, such as Rhett's disorder, mental retardation, and language disorders. All items are scored on a four-point Likert scale ranging from (0) "never observed" to (3) "frequently observed," meaning the individual behaves in this manner at least 5–6 times per 6-hour period. A parent or guardian will rate delays in social interactions and language and abnormal functioning in social interaction, language, and imaginative play during the first 3 years of the child's life. These "yes" or "no" questions are in accord with the diagnostic criteria for Autism from DSM-V. The GARS-3 coding system for videos was also used to ensure interrater reliability of facial expressions throughout the study.

Statistical Analysis

Data collected was analyzed using Microsoft Excel. Only the two normative children and the one ASD child were allowed to be video recorded during the study. Facial actions were coded for 10 second segments for the first and second iteration of the CPT, VAS pain rating scores were also graphed for the two iterations of the CPT. Only VAS pain rating scores were graphed for the other two normative children. The average VAS pain rating scores of the

normative children were then graphed alongside the VAS pain rating score of the ASD child. Similarly, the averages of the CFCS scores of the normative children were graphed alongside the CFCS score of the ASD child.

RESULTS

All participants were Caucasian males from the Capital region. Upon entering the lab, if the participant identified on the Autism Spectrum, the parent was administered the GARS-3 “Section 5: Ratings,” addressing “Restricted/Repetitive Behavior,” “Social Interaction,” “Social Communication,” “Emotional Responses,” “Cognitive Style,” and “Maladaptive Speech.” The mother of the one ASD child completed this form. There was a raw score of 10 in restricted/repetitive behaviors, a raw score of 12 in social interaction, a raw score of 10 in social communication, a raw score of 2 in emotional response, a raw score of 13 in cognitive style, and a raw score of 2 in maladaptive speech. These results coincide with a diagnosis of Asperger Syndrome.

For all the participants that consented, video recording was employed to later code facial actions. The graphs depict whether a facial action was present or not, not the specific facial action itself. For participant 1 during the first iteration of the CPT, he displayed distinct lip corner pulling from 20 seconds to 70 seconds, and at 60 seconds he blinked. During the second iteration, participant 1 had distinct lip corner pulling from 80 seconds to 110 seconds (see Figure 1). For participant 2 during the first iteration of the CPT, he displayed slight brow lowering at 40 seconds, and 60 seconds to 90 seconds. There was also slight lip corner pulling at 80 seconds, and slight horizontal mouth stretch from 70 seconds to 90 seconds (see Figure 2). For participant 5 (ASD child) during the first iteration of the CPT, he displayed slight lip corner pulling at 20 seconds, 50 seconds, and 80 seconds. He also showed distinct vertical mouth stretching at 40

seconds and 60 seconds. During the second iteration, this participant displayed the same results as during the first iteration (see Figure 5).

VAS pain ratings were also collected every 10 seconds from submersion of the child's hand into the CPT, until it was withdrawn. During the first iteration of the CPT, participant 1 noted onset of pain at 30 seconds with a VAS of 5, and withdrew his hand at 1-minute 12 seconds with a VAS of 9 (see Table 1). Participant 2 noted onset of pain at 10 seconds with a VAS of 7, and withdrew his hand at 2 minutes with a VAS of 7 (see Table 1). Participant 3 noted onset of pain at 20 seconds with a VAS of 3, and withdrew his hand at 1-minute 10 seconds with a VAS of 7 (see Table 1). Participant 4 noted onset of pain at 1-minute 5 seconds with a VAS of 5, and withdrew his hand at 2 minutes with a VAS of 6 (see Table 1). Lastly, participant 5 (ASD child) noted onset of pain at 40 seconds with a VAS of 4, and withdrew his hand at 1-minute 25 seconds with a VAS of 8 (see Table 2). VAS scores were also obtained for the same time points during the second iteration of the CPT. Participant 1 noted onset of pain at 30 seconds with a VAS of 6, and withdrew his hand at 1-minute 15 seconds with a VAS of 10 (see Table 1). Participant 2 noted onset of pain at 10 seconds with a VAS of 5, and withdrew his hand at 1-minute 40 seconds with a VAS of 8 (see Table 1). Participant 3 noted onset of pain at 20 seconds with a VAS of 2, and withdrew his hand at 1-minute 21 seconds with a VAS of 7 (see Table 1). Participant 4 noted onset of pain at 40 seconds with a VAS of 3, and withdrew his hand at 2 minutes with a VAS of 5 (see Table 1). Lastly, participant 5 (ASD child) noted onset of pain at 40 seconds with a VAS of 3, and withdrew his hand at 1-minute 39 seconds with a VAS of 8 (see Table 2).

A t-test between the two normative group children that had allowed video recording and the one ASD child for VAS pain ratings yielded a significant p-value ($p = 0.002$), suggesting the

normative group reported higher pain scores than the ASD child (see Figure 6). The average for the CFCS items were also taken for the two normative children over the two CPT iterations and compared against the CFCS over the two CPT iterations for the ASD child. This showed that all three children had the same number of facial features elicited during the CPT across the two iterations (see Figure 7).

Throughout the study, vitals were obtained at certain time points. Before the warm water bath, vitals i.e. blood pressure (mmHg) and pulse (beats/min) were obtained: Participant 1 had 131/75 and 60, participant 2 had 113/70 and 79, participant 3 had 110/90 and 76, participant 4 had 129/76 and 93, and participant 5 (ASD child) had 116/75 and 80. After the warm water bath and before the administration of the CPT, vitals were again obtained: Participant 1 had 135/74 and 65, participant 2 had 124/70 and 89, participant 3 had 123/96 and 92, participant 4 had 127/82 and 92, and participant 5 (ASD child) had 120/72 and 84. Following the CPT, the vitals were obtained for the last time during the first iteration of the study: Participant 1 had 117/74 and 65, participant 2 had 119/62 and 74, participant 3 had 118/93 and 84, participant 4 had 132/86 and 85, and participant 5 (ASD child) had 118/80 and 86. For the second iteration of the study the same vitals were taken at the same time points. Before the warm water bath: Participant 1 had 115/78 and 57, participant 2 had 115/71 and 78, participant 3 had 112/78 and 75, participant 4 had 130/76 and 83, and participant 5 (ASD child) had 122/76 and 82. After the warm water bath and before the CPT: Participant 1 had 130/66 and 62, participant 2 had 126/71 and 89, participant 3 had 122/88 and 87, participant 4 had 130/76 and 83, and participant 5 (ASD child) had 125/72 and 84. Finally, vitals were taken after the CPT: Participant 1 had 129/67 and 55, participant 2 had 126/74 and 73, participant 3 had 116/79 and 82, participant 4 had 125/72 and 86, and participant 5 (ASD child) had 122/82 and 78.

DISCUSSION

There is a recognizable disparity in the literature regarding the expression of pain in children on the Autism Spectrum compared to their normative peers. While Rattaz (2013) found that ASD children displayed slower recovery indicating higher stress levels, Duerden (2015) found that ASD children demonstrated decreased thermal sensitivity. The current research was conducted as a means to contribute to the current literature on the topic.

In the current research conducted, it was examined if a child on the Autism Spectrum expressed pain intensity similar to the normative population. It was hypothesized that children on the Autism Spectrum would be hypersensitive to pain or express the same amount of pain as the normative group. The hypothesis was not supported. Results indicated that the child on the Autism Spectrum subjectively expressed less pain, measured by lower VAS pain rating scores during the two CPT iterations as compared to his normative peers. However, there was only one ASD individual in this study, and could be non-representative of the entire ASD population as a whole.

An interesting finding was that while the ASD child reported lower VAS pain rating scores, when compared to the two normative children for facial action, both groups displayed the same number of facial actions over the two iterations of the CPT. This slightly coincides with the findings of Nader et al. (2004), however, Nader's study showed that ASD individuals had greater facial reactivity than their normative peers. It is also interesting to note that the blood pressure of all participants decreased after the CPT.

Implications

Based on the VAS scores, this research suggests that the child on the Autism Spectrum is hyposensitive to pain. If this statistic obtained proves true for other children on the Autism

Spectrum, there could be the possibility of chronic pain in this population as well as improper dosages of analgesic medications administered. However, the recruitment of only one ASD participant makes this anecdotal evidence.

Strengths

All of the participants in this study were tested in a designated room at Union College, with minimum auditory and visual stimuli. They were also tested with only the experimenter present in the room, and the experimenter maintained a neutral face for the duration of the study. While it has been shown that a mother's reaction during a CPT can affect the child's interpretation of pain, this was avoided by having the parent to stay in a different room for the duration of the study, allowing the parent to observe through a two-way mirror if desired.

While the four normative children arrived in the lab at the same time, it was ensured by another researcher that individuals who had already participated in the CPT did not disclose how long they had submerged their hands. In doing this, competitiveness was not fostered in the lab. Also, during the CPT, taking VAS pain rating scores every 10 seconds allowed for the development of a pain sensitivity range rather than just the pain threshold and withdrawal. While a self-report measure of pain via the VAS pain rating scale could have led to issues, vitals were also obtained along with facial actions through the CFCS. To enhance reliability of results, each participant went through two iterations of the study.

All of the participants in this study were also males. This allowed for gender-matching across the two groups. Therefore, differences are not able to be inferred based on another sex being present in the study.

Limitations

The most notable limitation of this study was the small sample size, especially with only one child to represent the ASD group. While all the children were white males, the normative group consisted of four 13-year-olds whereas the ASD group consisted of the one 16-year-old child. Therefore, the result that the ASD child expresses less pain than his normative peers could be an individual difference, or based on his ASD, the inability to properly perceive the VAS pain rating scale. Due to this, a single ASD case could be non-representative, and a future study with more ASD participants is necessary.

During the study, the CPT also did not have continuously circulating water. This would have aided in maintaining the necessary 10°C for the CPT, and preventing a possible microenvironment of warm water from forming around the child's hand. While the temperature on the thermometer was checked intermittently during the study, it is possible that some of the children were able to keep their hands submerged for a longer duration due to the formation of a warm microenvironment.

For each iteration of the study, the dominant hand was used for both the warm water bath and the CPT. There is a possible cumulative cooling effect associated with repeated immersion of the same hand, even following a warm water bath. In terms of the arm, the continual placement of the blood pressure cuff on the same arm may have impacted the readings throughout the study.

Future Research

An interesting facet to focus on for future research would be to assess the impact of anxiety, fatigue, and attention on pain thresholds for the participants, specifically the children on the Autism Spectrum. This could be instituted by distributing an inventory prior to the commencement of the study, providing participants with open ended questions for them to

answer about current anxiety level, fatigue state, and ability to pay attention. The ability for participants to elaborate would be particularly advantageous for ASD children since it has been shown that they are better able to accurately answer these types of questions as opposed to basing their feelings on a scale. If this type of study was conducted, I would expect that participants who noted that they were high in anxiety, high in fatigue, or lacked attention would display lower pain thresholds via higher scores on the VAS pain rating scale. It has been found that higher levels of stress and fatigue as well as lack of attention reduce the ability for an individual to modulate pain.

Another avenue to explore, specifically regarding the children on the Autism Spectrum would be the employment of a measure that notes an individual's fear of pain and catastrophizing. For example, it may be that an individual's fear of impending pain following the researcher's disclosure of what the CPT study entails, already makes the individual more susceptible to feel a greater amount of pain. The study would use the Fear of Pain Questionnaire (FPQ), which uses 30-items on a 5-point rating scale that measures fear regarding specific situations. The Pain Catastrophizing Scale (PCS) would also be administered, which measures rumination, magnification, and helplessness for 14-items on a 5-point scale. Lastly, the Coping Strategies Questionnaire (CSQ) would be used to measure pain coping strategies, through a 7-point scale to indicate the frequency with which a particular strategy is employed. Pain-related fear has been found to be a consistently stronger predictor of pain, more so than catastrophizing (Hirsh et al., 2008). It would be interesting to study if children on the Autism Spectrum have pain related fear, or if they may be unable to cognitively process painful stimuli enough to fear the pain stimulus prior to its introduction.

Conclusion

While there is a vast amount of research studying pain in children on the Autism Spectrum compared to their normative peers, varying methodology is used and different results have been found. This research added to the current literature by demonstrating that an ASD child, even if it is possibly an individual difference, expresses lower pain ratings yet the same amount of facial actions as the normative group. This could further support the assertion that ASD children may experience the same amount of pain, as evidenced by facial actions encoded by the CFCS, but they are unable to accurately express it, especially with a scale measure. Therefore, the limitations of this study should be focused on in successive studies to provide better insight into the pain experience of children on the Autism Spectrum.

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Figure 1. Normative pain ratings and facial actions for first and second iteration of the CPT

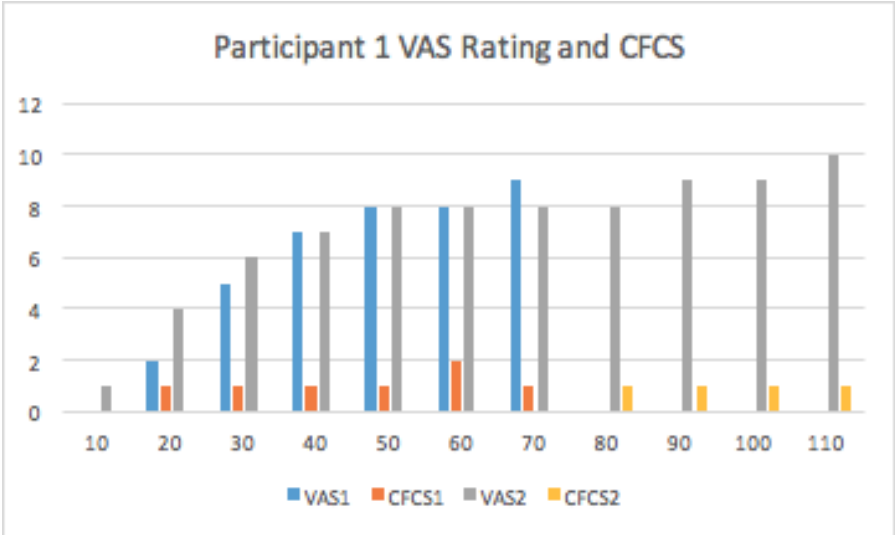


Figure 2. Normative pain ratings and facial actions for first and second iteration of the CPT

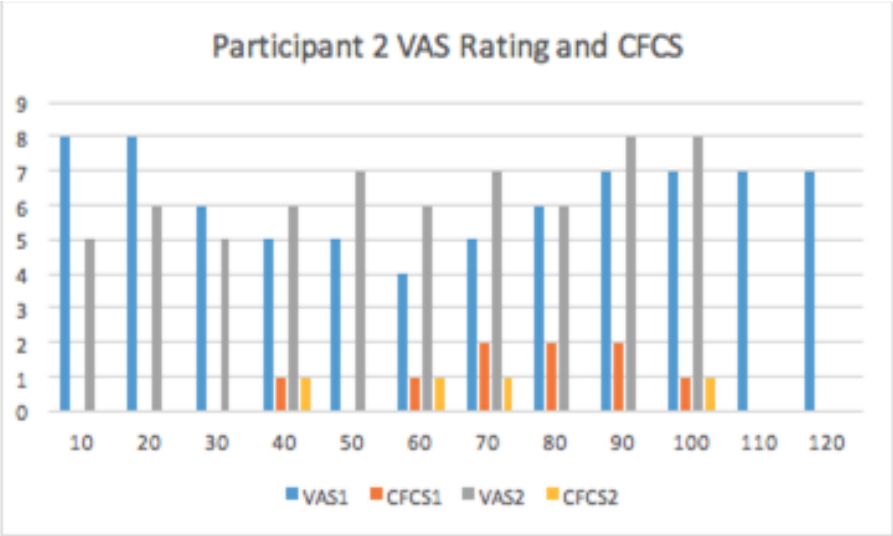


Figure 3. Normative pain ratings for first and second iteration of CPT

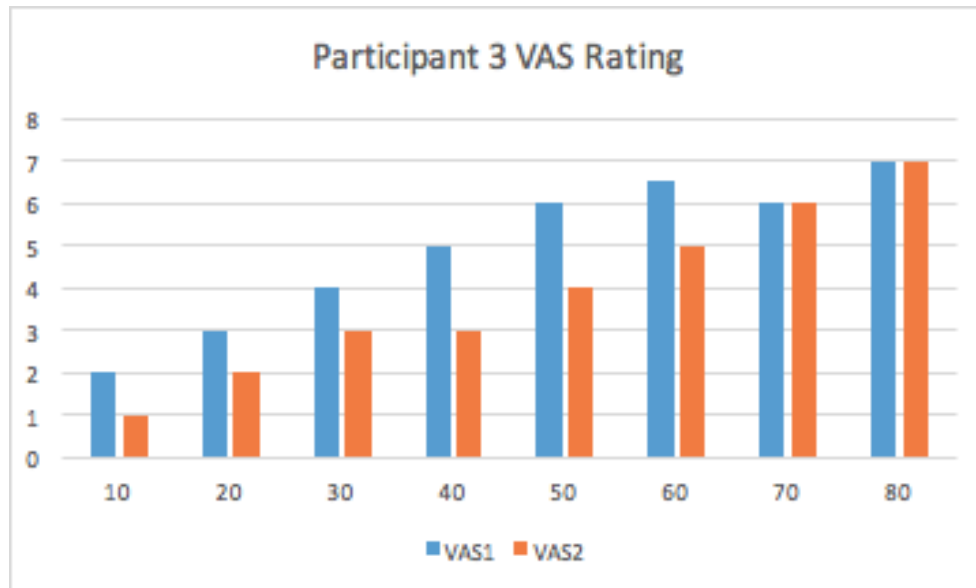


Figure 4. Normative pain ratings for first and second iteration of CPT

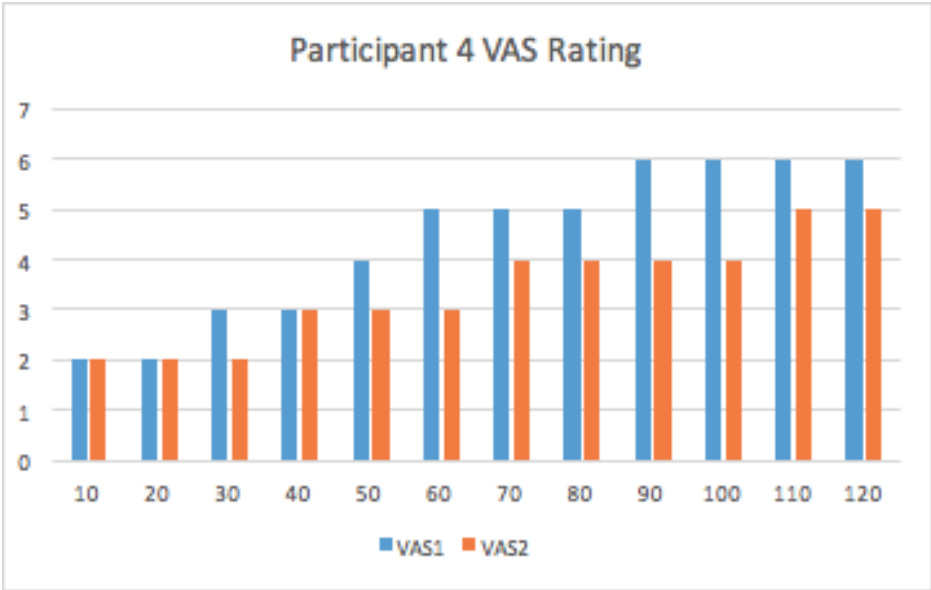


Figure 5. ASD pain ratings and facial actions for first and second iteration of the CPT

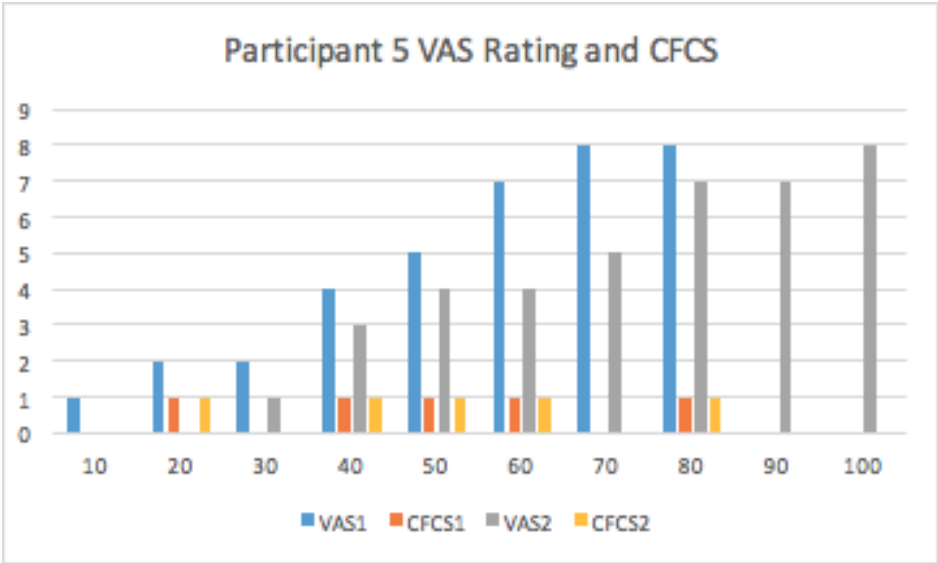


Figure 6. Normative pain ratings compared to ASD

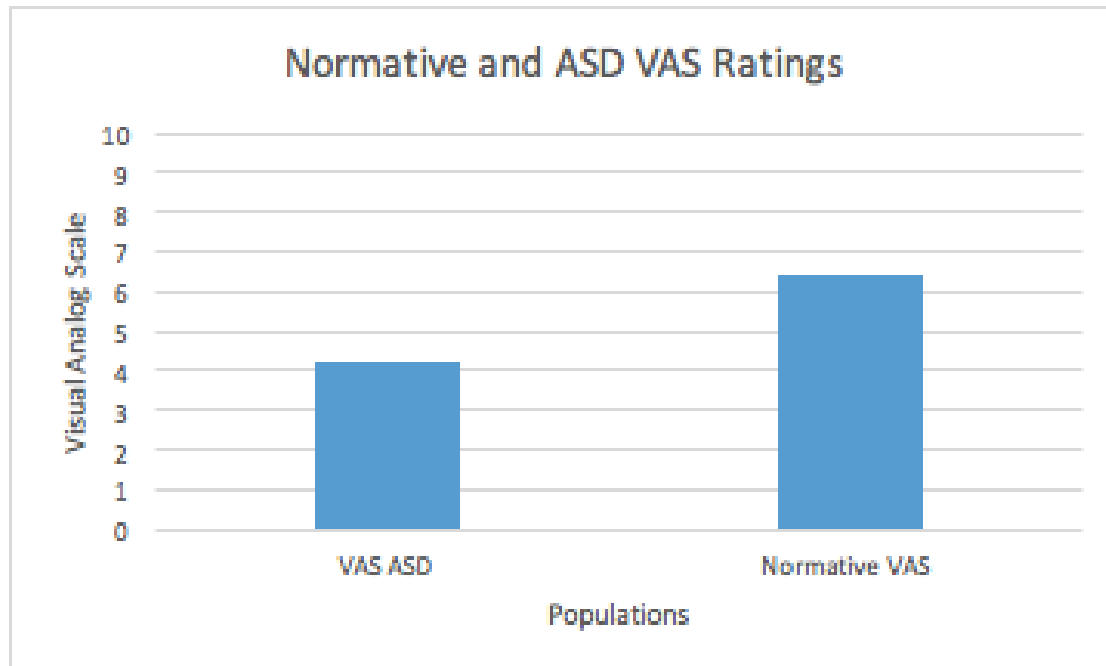


Figure 7. Normative facial actions compared to ASD

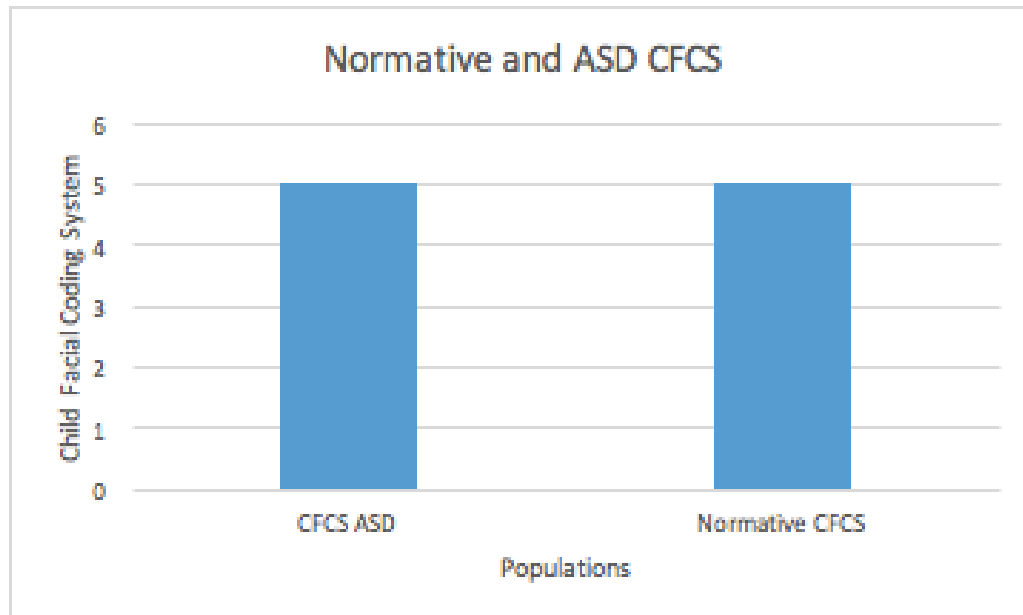


Table 1. Normative pain ratings for first and second iteration of CPT

VAS								
	Participant 1		Participant 2		Participant 3		Participant 4	
Time (s)	1 st CPT	2 nd CPT	1 st CPT	2 nd CPT	1 st CPT	2 nd CPT	1 st CPT	2 nd CPT
10	0	1	8	5	2	1	2	2
20	2	4	8	6	3	2	2	2
30	5	6	6	5	4	3	3	2
40	7	7	5	6	5	3	3	3
50	8	8	5	7	6	4	4	3
60	8	8	4	6	6.5	5	5	3
70	9	8	5	7	6	6	5	4
80		8	6	6	7	7	5	4
90		9	7	8			6	4
100		9	7	8			6	4
110		10	7				6	5
120			7				6	5

Table 2. ASD pain ratings for first and second iteration of CPT

VAS		
Time (s)	Participant 5	
	1 st CPT	2 nd CPT
10	1	0
20	2	0
30	2	1
40	4	3
50	5	4
60	7	4
70	8	5
80	8	7
90		7
100		8
110		
120		

APPENDIX A

Protocol Instructions

Participant ID# _____
Location: _____
Evaluator Initials _____

Date: _____
Time: _____

Instructions Form

_____ Welcome participant to the study.

I greatly appreciate you taking the time to meet with me today so that we might learn more about how pain is displayed in children. Please understand that most of what I say to you will be read directly from this packet in order to ensure consistency across evaluations. We want to make sure that the directions are explained to each participant in the same way to prevent any confusion. This evaluation process should take about 30 minutes. Please let me know if you have any questions at any time.

_____ Give parent and participant a copy of the Informed Consent Forms.

Before we begin, I would like to invite you (the parent/guardian) the opportunity to test the water that will be used in this study, and deem if you are comfortable with your child participating in this study.

Parent/guardian tests water temperatures. If parent/guardian is comfortable with the child participating proceed. If not, end study here.

If proceeding: Now, I'd like to start by going over some paperwork. Please read this Informed Consent form (give two sheets to parents, and one sheet to child) carefully and sign at the bottom (review consent with participant). If you have any questions, do not hesitate to ask.

_____ Administer Demographic Questionnaire and Gilliam Autism Rating Scale, 3rd edition

*Parents/Guardians will fill these out

Please fill out these questionnaires to the best of your ability. Remember that all answers will remain confidential.

_____ Take baseline vitals (blood pressure and pulse) and skin temperature (time to complete: 60 seconds, record)

Now I will take your blood pressure and heart rate by placing a blood pressure cuff on your upper arm. Good. Now I will take your skin temperature.

1st CPT:

Baseline BP: _____

Baseline Pulse: _____

2nd CPT:

Baseline BP: _____

Baseline Pulse: _____

_____ **Good. Now while standing, I want you to place your dominant, unclenched hand up to your wrist into the warm water bath for 2 minutes. I will tell you when the two minutes is done.**

*Demonstrate to the child how to submerge hand in water

_____ *Take skin temperature a second time as well as blood pressure and pulse*

Good. Now I will take your skin temperature again as well as your blood pressure and heart rate.

1st CPT:

2nd BP: _____

2nd Pulse: _____

2nd CPT:

2nd BP: _____

2nd Pulse: _____

____ Record VAS pain rating for onset of pain (pain threshold), 10 second intervals, and withdrawal of hand (pain tolerance), as well as the times based off the stopwatch for each of these. Complete the NCCPC for children on the Autism Spectrum at the pain threshold and pain tolerance. Complete the CFCS for both the ASD youth as well as the normative population. Videotaping throughout this procedure will be determined by parental consent, and if allowed, myself and my lab assistant will independently rate facial pain.

*There is a 2 minute max hand submersion in cold water

Now, please place your same dominant, unclenched hand up to your wrist into the cold water bath. Please tell me when you first feel the pain, and I will ask for a pain rating. I will then ask you to give a pain rating on a scale of 0 to 10 (0 being no pain, and 10 being unbearable pain) every 10 seconds during the trial. When you withdraw your hand, I will ask you again to give me a pain rating. Please try and keep your hand in the water until it becomes painful, however, you can remove your hand at any time.

1st CPT:

1)____ 2)____ 3)____ 4)____ 5)____ 6)____ 7)____ 8)____ 9)____ 10)____

11)____ 12)____

Onset of pain (pain threshold):____ **Withdrawal (pain tolerance):**_____

2nd CPT:

1)____ 2)____ 3)____ 4)____ 5)____ 6)____ 7)____ 8)____ 9)____ 10)____

11)____ 12)____

Onset of pain (pain threshold):____ **Withdrawal (pain tolerance):**_____

____ Take vitals (time to complete: 60 seconds, record)

Good. Now I will take your heart rate and blood pressure again.

1st CPT:

3rd BP:_____

3rd Pulse:_____

2nd CPT:

3rd BP:_____

3rd Pulse:_____

____ **You will be given a 2 minute rest period, and then will be asked to complete the heart rate, blood pressure, skin temperature, warm water, and then cold water bath with pain ratings again.**

(Part B)*Video system will be employed throughout if allowed by parent, and will be coded by myself and another rater using GARS-3 guidelines.

____ Debrief, compensate, and thank participant.

APPENDIX B

Demographic Questions

ID#: _____

Date: _____

Years of Education (High School = 12)

First language = (English or list other?)

Gender (male or female?)

Which hand do you write with?

Which hand do you use to throw a ball?

Ethnicity (circle as many as apply)

Caucasian/White

African-American/Black

Hispanic-American

Asian-American

Native American

Other: _____

Age _____

Weight (lbs.): _____

Height (ft/in): _____

*GARS-3 Questionnaire will be administered here as well

Materials

xlii

GARS-3 – Parent Interview

Section 5: Ratings

Directions: On a scale of 0 to 3, rate the following items in terms of how adequately the item describes the individual's behavior. Circle the number that best describes your observations of the person's typical behavior under ordinary circumstances (i.e., in most places, with people he or she is familiar with, and in usual daily activities). Remember to rate every item. If you are uncertain about how to rate an item, delay the rating and observe the person for a 6-hour period to determine your rating.

0 Not at all like the individual
1 Not much like the individual
2 Somewhat like the individual
3 Very much like the individual

PLEASE RATE EVERY ITEM

Restricted/Repetitive Behaviors

1. If left alone, the majority of the individual's time will be spent in repetitive or stereotyped behaviors.	0	1	2	3
2. Is preoccupied with specific stimuli that are abnormal in intensity.	0	1	2	3
3. Stares at hands, objects, or items in the environment for at least 5 seconds.	0	1	2	3
4. Flicks fingers rapidly in front of eyes for periods of 5 seconds or more.	0	1	2	3
5. Makes rapid lunging, darting movements when moving from place to place.	0	1	2	3
6. Flaps hands or fingers in front of face or at sides.	0	1	2	3
7. Makes high-pitched sounds (e.g., eye-see-see-see) or other vocalizations for self-stimulation.	0	1	2	3
8. Uses toys or objects inappropriately (e.g., spins cars, takes action toys apart).	0	1	2	3
9. Does certain things repetitively, ritualistically.	0	1	2	3
10. Engages in stereotyped behaviors when playing with toys or objects.	0	1	2	3
11. Repeats unintelligible sounds (babbles) over and over.	0	1	2	3
12. Shows unusual interest in sensory aspects of play materials, body parts, or objects.	0	1	2	3
13. Displays ritualistic or compulsive behaviors.	0	1	2	3
Subtotals	+	+	+	

Restricted/Repetitive Behaviors Raw Score

Social Interaction

14. Does not initiate conversations with peers or others.	0	1	2	3
15. Pays little or no attention to what peers are doing.	0	1	2	3
16. Fails to imitate other people in games or learning activities.	0	1	2	3
17. Doesn't follow other's gestures (cues) to look at something (e.g., when other person nods head, points, or uses other body language cues).	0	1	2	3
18. Seems indifferent to other person's attention (doesn't try to get, maintain, or direct the other person's attention).	0	1	2	3
19. Shows minimal expressed pleasure when interacting with others.	0	1	2	3
20. Displays little or no excitement in showing toys or objects to others.	0	1	2	3
21. Seems uninterested in pointing out things in the environment to others.	0	1	2	3
22. Seems unwilling or reluctant to get others to interact with him or her.	0	1	2	3
23. Shows minimal or no response when others attempt to interact with him or her.	0	1	2	3
24. Displays little or no reciprocal social communication (e.g., doesn't voluntarily say "bye-bye" in response to another person saying "bye-bye" to him or her).	0	1	2	3
25. Doesn't try to make friends with other people.	0	1	2	3
26. Fails to engage in creative, imaginative play.	0	1	2	3
27. Shows little or no interest in other people.	0	1	2	3
Subtotals	+	+	+	

Social Interaction Raw Score

Social Communication

28. Responds inappropriately to humorous stimuli (e.g., doesn't laugh at jokes, cartoons, funny stories).	0	1	2	3
29. Has difficulty understanding jokes.	0	1	2	3
30. Has difficulty understanding slang expressions.	0	1	2	3
31. Has difficulty identifying when someone is teasing.	0	1	2	3
32. Has difficulty understanding when he or she is being ridiculed.	0	1	2	3
33. Has difficulty understanding what causes people to dislike him or her.	0	1	2	3
34. Fails to predict probable consequences in social events.	0	1	2	3
35. Doesn't seem to understand that people have thoughts and feelings different from his or her.	0	1	2	3
36. Doesn't seem to understand that the other person doesn't know something.	0	1	2	3
Subtotals	+	+	+	

Social Communication Raw Score

Emotional Responses

37. Needs an excessive amount of reassurance if things are changed or go wrong.	0	1	2	3
38. Becomes frustrated quickly when he or she cannot do something.	0	1	2	3
39. Temper tantrums when frustrated.	0	1	2	3
40. Becomes upset when routines are changed.	0	1	2	3
41. Responds negatively when given commands, requests, or directions.	0	1	2	3
42. Has extreme reactions (e.g., cries, screams, tantrums) in response to loud, unexpected noise.	0	1	2	3
43. Temper tantrums when doesn't get his or her way.	0	1	2	3
44. Temper tantrums when told to stop doing something he or she enjoys doing.	0	1	2	3
Subtotals	+	+	+	

Emotional Responses Raw Score

Is the individual mute? Yes No If your answer is yes, do not complete the next two subscales.

Cognitive Style

45. Uses exceptionally precise speech.	0	1	2	3
46. Attaches very concrete meanings to words.	0	1	2	3
47. Talks about a single subject excessively.	0	1	2	3
48. Displays superior knowledge or skill in specific subjects.	0	1	2	3
49. Displays excellent memory.	0	1	2	3
50. Shows an intense, obsessive interest in specific intellectual subjects.	0	1	2	3
51. Makes naive remarks (unaware of reaction produced in others).	0	1	2	3
Subtotals	+	+	+	

Cognitive Style Raw Score

Maladaptive Speech

52. Repeats (echoes) words or phrases verbally or with signs.	0	1	2	3
53. Repeats words out of context (repeats words or phrases heard at an earlier time).	0	1	2	3
54. Speaks (or signs) with flat tone, affect.	0	1	2	3
55. Uses "yes" and "no" inappropriately. Says "yes" when asked if he or she wants an aversive stimulus or says "no" when asked if he or she wants a favorite toy or treat.	0	1	2	3
56. Uses "he" or "she" instead of "I" when referring to self.	0	1	2	3
57. Speech is abnormal in tone, volume, or rate.	0	1	2	3
58. Utters idiosyncratic words or phrases that have no meaning to others.	0	1	2	3
Subtotals	+	+	+	

Maladaptive Speech Raw Score

The CPT Setup:

